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Awareness of sudden unexpected death in epilepsy among parents of children with epilepsy in a tertiary center

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ABSTRACT

Objective: Sudden unexpected death in epilepsy (SUDEP) is the second leading neurological cause of potential years of lifelost after stroke. Despite growing numbers of studies on social aspects of epilepsy, there is a paucity of research on the awareness of SUDEP among the parents of children with epilepsy (CWE), especially in Turkey. In this study, we aimed to evaluate the knowledge of parents of the CWE regarding SUDEP in the tertiary setting.
Material and methods: A total of 146 parents (108 female) aged 19 to 55 years (median age:34) of CWE were included at Marmara University, School of Medicine, Department of Pediatric Neurology outpatient clinic between May 2018 and September 2018. A total of 30 multiple-choice questions and a written survey were administered, which consisted of three sections. In the first section, the sociodemographics of parents and CWE were questioned. In the second section, the severity of epilepsy was evaluated. In the third section, the knowledge level and awareness of parents of CWE were assessed.
Results: Of 146 parents, only 16.6% previously heard about SUDEP, while 45% of them heard from their relatives. The presence of prior knowledge of SUDEP was associated with the presence of prolonged postictal confusion and longer duration of epilepsy ($p < 0.05$). Ninety-seven (66%) parents desired to be informed about SUDEP, while 76 (54.7%) of them agreed that this information should be given at the time of diagnosis. The degree of anxiety in parents regarding death of epilepsy-related causes was significantly related with prolonged postictal confusion ($p < 0.001$) and using three or more antiepileptic drugs ($p = 0.005$).
Conclusion: Our data suggest that knowledge about SUDEP among parents with CWE found inadequate in Turkey. There should be much effort to inform parents and caregivers in epilepsy clinics on SUDEP, which may help to reduce the associated risk factors.

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1. Introduction

Epilepsy is one of the most common and disabling neurological conditions worldwide. Sudden unexpected death in epilepsy (SUDEP) is explicitly defined as the sudden, unexpected, witnessed or unwitnessed, nontraumatic and nondrowning death of patients with epilepsy (PWE) with or without evidence of a seizure, and excluding documented status epilepticus in whom postmortem studies reveal no structural or toxicological cause for death [1]. It is the second leading neurological cause of potential life-year lost after stroke. In the United Kingdom, about half of 1000 epilepsy-related deaths are documented due to SUDEP each year [2]. According to the epidemiological studies, the risk for SUDEP in children with epilepsy (CWE) aged between 0 and 17 years is 0.22/1000 patient-years and the risk increases in adults to 1.2/1000 patient-years [3]. However, in recent studies, SUDEP has been more frequently reported in children than before [4], a nationwide population-based cohort study reported similar rates of SUDEP in children and adults [5]. Moreover in low- and middle-income countries there is a lack of information about the incidence of SUDEP [6]. The main restriction for determination of SUDEP is the challenges in identification of an accurate cause of death with limited resources for a postmortem investigation [7]. Furthermore, the refusal of an autopsy in sudden death secondary to family wishes and cultural considerations precludes real numbers about the incidence of SUDEP [8].

The lifetime cumulative risk for SUDEP by the age of 40 has been estimated at 7% overall, or 12% in those with persistent epilepsy [9]. There are few factors that have been shown to be associated with an increased risk for SUDEP in adults: the presence of generalized tonic–clonic seizures (GTCS), having more than three GTCS per year, the absence of seizure freedom for 1- to 5-year intervals, long duration of epilepsy of over 15 years and nocturnal seizures without surveillance. On the other hand,

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Questionnaire Section 1

Q1-6. Asks for respondents' age, sex, highest education level, income status and working status. Responses are included in Table 1.

Q7-9. Asks for CWE's age and sex. Responses are included in Table 1.

Questionnaire Section 2

Q10. How old was your child when the seizures began?

Q11. How many medications are your child currently taking to control seizures?

Q12. Did your child have seizures within the past year? (Answers: Yes, No)

Q13. How long of your child's post-ictal confusion? (Answers: mild: less than 10 minutes, moderate: 10 minutes to 1 hour, severe: more than 1 hour)

Q14. How often do your child had seizures in the last year?

Q15. How many severe seizures have your child ever had in his/her life? (Answers: 0-5, ≥6, Don't know)

Q16. Have your child ever had a seizure lasting more than 5 to 10 min? (Answers: Yes, No, Don't know)

Q17. Have you referred to the ED due to seizure within the past year? (Answers: Yes, No)

Q18. Does your child regularly attend to the outpatient follow-up visit scheduled by the treating physician as recommended? (Answers: On a regular basis, Not regularly, If necessary)

Questionnaire Section 3

Q19. Have you heard of SUDEP (Sudden Unexpected Death in Epilepsy)? (Answers: Yes, No)

Questionnaire Section 3A

Q20. How did you first learn about SUDEP? (Answers: Doctor, Friend/Relative, Internet or Media)

Q21. Do you feel well-educated about SUDEP? (Answers: Yes, No, Probably)

Q22. Do you think your child at risk for SUDEP? (Answers: Yes, No, Don't know)

Q23. When you first heard about SUDEP, how did you feel? (Answers: Scared, Frustrated, Annoyed, Confused)

Q24. If you are willing to obtain more information about SUDEP, from whom do you get? (Answers: Doctor, Others)

Q25. Is there any way to prevent SUDEP in your child? (Answers: Using AED regularly, Using audible and visual display to monitor my child's seizures, Encouraging my child to exercise regularly, Avoiding behaviors which trigger the epileptic seizure of my child, Adopting a regular sleeping schedule, Others)

Questionnaire Section 3B

Q26: How much do you worry about your child dying from epilepsy or seizures? (Answers: Too much, a little, not at all)

Q27. What can cause epilepsy-related death in your child? (Answers: Accident, Drowning, Seizures more than 5 to 10 min, SUDEP)

Q28. Do you want more information about SUDEP? (Answers: No, I don't / Yes, if my child is at high risk for SUDEP / Yes, regardless of the existing risk)

Q29. If your medical provider would discuss SUDEP with you, when would it be the best time for the first talk about it? (Answers: When I ask for information / If my child's seizures are not under control / During a follow-up visit, regardless of current seizure control / Immediately before initiating anti-seizure medications / When my child was first diagnosed with epilepsy / Other)

Q30. Do you think that learning more about SUDEP could make a difference in how well you take care of your child's health, including using AEDs regularly? (Answers: Yes, No, Probably)

Q31. Which one of the following options can reduce your child's risk for epilepsy-related death? (Answers: Using AEDs regularly, Using audible and visual display to monitor seizures, while my child is sleeping at night, Encouraging my child to exercise regularly, Avoiding behaviors which trigger the epileptic seizure of my child, Adopting a regular sleeping schedule, Others)

Fig. 1. A 30-multiple-choice questionnaire consisting of three sections in the English language (translated version). SUDEP, Sudden unexpected death in epilepsy; CWE, children with epilepsy; AED, antiepileptic drug; ED, emergency department.

improved or full seizure control by effective adjunctive antiepileptic treatment has been demonstrated to reduce the risk for SUDEP [10,11]. These potentially modifiable risk factors can be used as a part of structured risk assessment and information giving in adults [12]. Nevertheless, risk factors for SUDEP in children are not completely understood. A recent review classified factors influencing SUDEP risk in children as etiology-related, environmental, seizure, treatment, and other. Seizure factors were subgrouped as seizure frequency, severity, uncontrolled seizures, and failure of 5-year terminal remission [13]. Of note, adolescents are more vulnerable to increase SUDEP risk due to nonadherence to antiepileptic drugs (AEDs), sleep deprivation, and increased risk-taking behavior with use of alcohol.

Recently, consensus guidelines state that the potentially increased risk for death associated with epilepsy should be defined as a part of the comprehensive epilepsy education provided to PWE and parents with CWE [14,15]. However, these guidelines have not been fully implemented by

treating physicians in every country [16–21]. Despite growing numbers of studies on PWE, there is still a limited number of studies regarding the awareness of SUDEP among the parents of CWE. In a survey including 67 parents of CWE, 91% of parents were willing to be informed about SUDEP, and 67% desired to be informed at the time of diagnosis, while only 3% preferred not to be informed more specifically [17]. These findings were also supported in a qualitative study including parents of CWE, which reported that the majority of the parents preferred to be informed about SUDEP more clearly [22]. There are also several studies showing that the majority of parents of CWE prefer SUDEP counseling at the time of diagnosis of epilepsy [23–25].

Furthermore, discussion of SUDEP with parents and caregivers early in the clinical course of the disease is paramount, which may allow to change behavior or take preventive measures. In the present study, we, therefore, aimed to evaluate the knowledge of parents of CWE regarding SUDEP in the tertiary setting as a first survey done in Turkey.

2. Material and methods

2.1. Study design and sample

This descriptive, single-center study was carried out at Marmara University, School of Medicine, Department of Pediatric Neurology outpatient clinic between May 2018 and September 2018 and included parents of children with an established diagnosis of epilepsy. Children's age admitted to the pediatric neurology outpatient clinic ranged from newborns up to 22 years olds. Our epilepsy clinic supervised by the two senior epileptologists has accepted all pediatric patients with epilepsy, ranging from self-limited epilepsies to epileptic encephalopathies and also is a referral center for medically resistant patients with epilepsy to be evaluated for alternative therapy options as epilepsy surgery, vagus nerve stimulation, or ketogenic diet. The study protocol was approved by the local Ethics Committee (No: 09.2018.262, Date: 06.04.2018).

Some of the parents of CWE included in the study were on regular follow-up, some of them were newly-admitted to the clinic. Exclusion criteria were as follows: having a single unprovoked seizure, structural brain deformities, and active discontinuation of treatment. We included parents of CWE with nonlesional epilepsy to assess their knowledge only about sudden unexpected death related with epilepsy. The presence of structural lesions, especially severe lesions of the central nervous system like hydrocephalus may stimulate them to search for morbidity and mortality related with the lesions and also for surgical consequences of these pathologies.

All parents were informed about the nature of the study and seven refused to give their consent. Finally, a total of 146 parents of CWE were included in the study. A written informed consent was obtained from each parent. Each parent of CWE who was first seen in clinic-setting was consented in person. In order to evaluate the awareness and knowledge of the main caregiver of the CWE, even when two of the parents were in the clinic for follow-up visit; we asked the main caregiver to have the interview. We considered the opinion of the caregiver, regardless of the other parent's education level or interest in epilepsy-related death.

Parents who were not aware of SUDEP and wanted to have more information were directed to their follow-up epileptologists whom consented and agreed to give accurate information by using definitions based on the latest literature. Attended epileptologist also informed parents who had previous knowledge about SUDEP upon request. However, discussion on SUDEP has not been a part of regular follow-up yet.

The study was conducted in accordance with the principles of the Declaration of Helsinki.

2.2. Data collection

A questionnaire of a 31-multiple-choice questions and a written survey were used for the data collection. Each parent of CWE was consented in person by two intern physicians (D.A. and S.B.K.) under the supervision of the pediatric neurologist (D.T.). A single survey was administered to each parent by reading all questions explicitly in Turkish during face-to-face interviews. The true translation of the questionnaire into the English language is shown in Fig. 1.

2.3. Questionnaire

The questionnaire consisted of three sections. In the first section, the sociodemographics of parents and CWE including age, sex, education level, income status, and working status were questioned. In the second section, the characteristics of epilepsy and seizures were evaluated. Parents of CWE were asked about seizure onset (age), duration of epilepsy (years), current number of AEDs, having a seizure in the past year, frequency of seizures, duration of postictal confusion, lifetime number of

Table 1
Demographics of parents and CWE and characteristics of seizures.

		n	%
Sex	Female	108	74.0
	Male	38	26.0
Age, years, median (min-max) 34 (19–55)*			
Highest education level	Primary school or less	67	46.5
	Secondary school	39	27.1
	High school, college, and postgraduate	38	26.4
Income status	Expenses are more than my revenue	19	14.5
	Expenses are equal to my revenue	73	55.7
	Expenses are less than my revenue	39	29.8
Working status	Unemployed	92	63.9
	Employed	52	36.1
Age of CWE, years, mean ± SD 7.73 ± SD 5.01			
Sex of CWE	Female	80	54.8
	Male	66	45.2
Seizure onset age, mean ± SD 3.2 ± SD 3.495**			
Duration of epilepsy, years	0–3	80	55.2
	≥4	65	44.8
Duration of epilepsy, years, median 3 (0–18)*			
Current number of AEDs	0	3	2.1
	1–2	117	80.7
	≥3	25	17.2
Seizures within the past year	No	39	26.7
	Yes	107	73.7
Frequency of seizures	More than one seizure a day	5	3.4
	More than two seizures per month	19	13.0
	10–20 seizures per year	29	19.9
	1–9 seizures per year	54	37.0
	No seizures within the past year	39	26.7
Severity of post-ictal confusion	Mild	52	36.1
	Moderate	40	27.8
	Severe	52	36.1
Lifetime number of GTCS	0–5	83	58.5
	≥6	28	19.7
	Don't know	31	21.8
Seizure lasting more than 5 to 10 min	Yes	56	38.4
	No	66	45.2
	Don't know	24	16.4
Admission to ED due to a seizure within the past year	Yes	63	43.8
	No	81	56.2
Outpatient follow-up status	On a regular basis	126	86.3
	Not regularly	18	12.3
	If necessary	2	1.4

Data are given in number and percentage, unless otherwise stated.* The median (min-max) values are given, since the participants' age and duration of epilepsy constant variables do not fit the normal distribution.** The mean ± SD is given for CWE's age and seizure onset age, since these variables fit the normal distribution. SUDEP, Sudden unexpected death in epilepsy; CWE, children with epilepsy; AED, antiepileptic drug; ED, emergency department; SD, standard deviation; GTCS, generalized tonic–clonic seizure.

GTCS, seizure lasting more than 5–10 min, admission to emergency department due to a seizure past year and outpatient follow-up status. Seizure frequency was classified as 1 to 9 seizures per year or 10–20 seizures per year occurring during actual year or more than 2 seizures per month or more than 1 seizure per day occurring during the recent 3 months. Answers given to duration of postictal confusion (Q13) were stratified into 3 subgroups as mild (less than 10 min), moderate (10 to 60 min) or severe (longer than 60 min).

In the third section of the questionnaire, the parents of CWE were asked whether they had heard of SUDEP previously in the 3A section. Among respondents who never heard of SUDEP before were asked to skip to the 3B section. For respondents who heard of SUDEP before were asked to fill the part 3A, which evaluated how they first heard of it and whether they felt that their child was at risk for SUDEP, and asked to identify their reasons. In the 3B section, the respondents were asked about their opinions and attitudes toward epilepsy-related death events and the way of coping with it. The degree of anxiety and postictal confusion was assessed using a three-point Likert-type rating scale: Too much/severe, a little/moderate, not at all/mild.

2.4. Statistical analysis

Statistical analysis was performed using the Statistical Package for the Social Sciences (SPSS) version 25.0 software (IBM Corp., Armonk, NY, USA). Descriptive data were expressed in mean ± standard deviation (SD), median (min-max), or number and frequency. The Pearson chi-square or Fisher's exact tests were used to analyze the significance of differences between the groups of interest. A p value of <0.05 was considered statistically significant.

For all participants, frequency analysis was done for demographics of parents and CWE and characteristics of their epilepsy are shown in Table 1.

3. Results

3.1. Characteristics of study population

Out of 146 participants, 108 (73%) were females with a median age of 34 (range, 19 to 55) years. Sixty-seven (46.5%) parents graduated from primary school or less and only 38 (26.4%) of them graduated from high school and above (high school, college, or postgraduate). Ninety-two (63.9%) parents were unemployed, and 39 (29.8%) parents stated that their expenses were less than their revenue.

About the characteristics of CWE, eighty CWE (54.8%) were female, and the mean age of the CWE was 7.73 ± 5013. The number of children who had been with epilepsy diagnosis for more than 4 years was 65 (44.8%), and 80 (55.2%) patients had the diagnosis for 0–3 years. Twenty-five (17.2%) CWE used 3 or more AEDs, and 107 (73.7%) of the CWE had seizures in the previous year. Of them, 52 (36.1%) children had severe ictal and postictal confusion, and 28 (19.7%) children had had 6 or more generalized tonic–clonic seizures in their lifetime. According to the observations of the parents, 56 (38.4%) CWE had at least one generalized seizure that lasts more than 10 min. One hundred twenty-six (86.3%) of the parents stated that there were on a regular outpatient clinic follow-up for their CWE. Demographic characteristics of parents and CWE and characteristics of seizures are shown in Table 1.

3.2. Prior knowledge of SUDEP

Out of 146 parents, 122 (83%) never heard of SUDEP before and 24 (16%) had prior knowledge of SUDEP. Eleven (45.8%) parents who heard of SUDEP before initially learned from their friends or relatives, while 9 (37.5%) parents were informed by the treating physician and only 4 (16.7%) of them learned from online resources. At the first the participants heard of SUDEP, 16 (69.6%) were scared and worried. Thirteen (56.5%) of the parents believed that their children were at risk for SUDEP, while 10 (41.7%) believed that their knowledge of SUDEP was sufficient. In addition, 20 (87%) of the parents preferred visiting their treating physician to obtain more information about SUDEP. The parents who had prior knowledge of SUDEP believed that the following precautions would be preventive for SUDEP: regular use of AEDs (47.1%), avoiding their children's epileptic seizure-triggering behaviors (19.6%), using audible and visual display to monitor the child's seizures (13.7%), and adopting a regular sleeping schedule of the child (9.8%). Three parents use other methods to prevent SUDEP in their children, such as, having physiotherapy, chewing and swallowing therapy for their child, controlling their child not to have high fever, and preventing their child from vomiting (Fig. 2).

3.3. Parents' perception on epilepsy-related death and SUDEP

Of all parents, 52 (37%) were worried about the death of their child due to an epilepsy-related cause. Ninety-seven (66%) parents were willing to obtain more information about SUDEP, irrespective of the existing risk for SUDEP. Seventy-six (54%) parents preferred to obtain information at the time of the initial diagnosis. Among the respondents, 90 (64.3%) believed that a more detailed information about SUDEP could make a difference in improving their child's health, such as using AEDs on a regular basis (Table 2).

Based on the characteristics of epilepsy of the child and parents' awareness about SUDEP, prior knowledge of SUDEP was significantly found to be related with prolonged postictal confusion ($p = 0.007$), seizures lasting more than 5 min ($p < 0.001$), using three or more AEDs ($p < 0.001$), and lifetime history of having ≥6 GTCS ($p < 0.001$). However, there was no significant association between frequency of seizures and knowledge of SUDEP of the parents (Table 3).

The degree of anxiety about death of epilepsy-related cause among parents was significantly related with prolonged postictal confusion (p

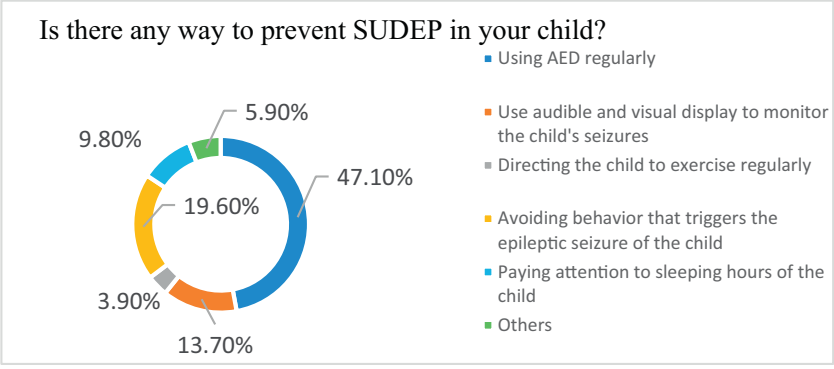


Fig. 2. Parents' preferred methods to prevent SUDEP in their child SUDEP, Sudden unexpected death in epilepsy; AED, antiepileptic drug.

Table 2
Parents' perception on epilepsy-related death and SUDEP.

		n	%
Do you worry about your child to have an epilepsy-related death?	Not at all	65	46.4
	A little	23	16.4
	Too much	52	37.1
What can cause the epilepsy-related death in your child?	Accident	42	19.4
	Drowning	59	27.2
	Seizure lasting more than 5 to 10 min	83	38.2
	SUDEP	33	15.2
	No, I don't	14	9.7
Do you want more information about SUDEP?	Yes, if my child is at high risk for SUDEP	34	23.4
	Yes, regardless of the existing risk	97	66.9
If your medical provider would discuss SUDEP with your, when would it be the best time for the first talk about it?	When I ask for information	14	10.1
	If my child's seizures are not under control	19	13.7
	During a follow-up visit, regardless of current seizure control	12	8.6
	Immediately before initiating anti-seizure medications	17	12.2
	When my child was first diagnosed with epilepsy	76	54.7
	Other	1	0.7
Do you think that learning more about SUDEP could make a difference in how well you take care of your child's health, including using AEDs regularly?	Yes	90	64.3
	No	19	13.6
	Maybe	31	22.1

Data are given in number and percentage, unless otherwise stated. SUDEP, Sudden unexpected death in epilepsy; AED, antiepileptic drug.

Table 3
Comparison of epilepsy characteristics and prior knowledge of SUDEP.

		Heard of SUDEP before				
		No		Yes		
		n	%	n	%	
Severity of postictal confusion	Mild	45	37.5	7	29.2	p =
	Moderate	38	31.7	2	8.3	0.007
	Severe	37	30.8	15	62.5	
Seizure lasting more than 5 to 10 min	Yes	38	31.1	18	75.0	p <
	No	61	50.0	5	20.8	0,001
	Don't know	23	18.9	1	4.2	
Current number of AEDs	0–2	109	90.1	11	45.9	p <
	≥3	12	9.9	13	54.2	0,001
Lifetime number of GTCS	0–5	77	64.7	6	26.1	p <
	≥6	17	14.3	11	47.8	0,001
	Don't know	25	21.0	6	26.1	
History of seizures within the past year	Yes	89	73.6	18	75.0	p =
	No	32	26.4	6	25.0	0,883
Frequency of seizures	More than one seizure a day	4	4.8	1	4.1	p = 0,285
	More than two seizures per month	6	7.2	13	54.1	
	10–20 seizures per year	23	27.7	6	25.0	
	1–9 seizures per year	50	60.2	4	16.6	
Highest education level	Primary school or less	56	46.3	11	47.8	p = 0,158
	Secondary school	36	29.8	3	13.0	
	High school, college, and postgraduate	29	24.0	9	39.1	
Do you want more information about SUDEP?	No, I don't	14	11.6	0	.0	p =
	Yes, if my child is at high risk for SUDEP	28	23.1	6	25.0	0,213
	Yes, regardless of the existing risk	79	65.3	18	75.0	

Data are given in number and percentage, unless otherwise stated. SUDEP, Sudden unexpected death in epilepsy; AED, antiepileptic drug; GTCS, generalized tonic–clonic seizure.

< 0.001), lifetime history of having ≥6 GTCS (p = 0.011), and using three or more AEDs (p = 0.005), but not significantly associated with the duration of epilepsy, having seizures within the past year or frequency of seizures (Table 4).

4. Discussion

This study is a hospital-based, face-to-face survey including 146 parents of CWE. To the best of our knowledge, this is the first large-scale study on this topic. Being the most populated city of Turkey, our outpatient clinic in Istanbul province has a remarkable spectrum of PWE from different regions of the country, and thus, a large number of participants was included in the study to obtain relatively robust findings.

Based on our study results, only a very limited number of the parents heard about SUDEP before, which is relatively low compared to the overall limited data on parental knowledge about SUDEP in the literature. In previous studies in different countries, the degree of awareness of SUDEP among parents of CWE varied between 31.5 and 34.4% including moderate-to-high socioeconomic status of the population. The discrepancy between our study and previous studies can be attributed to the lower socioeconomic status of our study sample. A minority of our parents graduated from a university compared to other studies; thus, the ratio was much higher [24,26], and also 63.9% of our parents currently do not have any jobs.

Furthermore, in the present study, nearly half of the parents who had existing knowledge on SUDEP obtained this information from their friends or relatives, while only one-third of the parents learned from their treating physicians. In their study, Fong et al. [24] reported that Malaysian parents who had already heard of SUDEP before mostly obtained this information from hospital doctors. In our study group, the information about SUDEP was not initially obtained from treating physicians; instead, they gained more information from their relatives or friends. This difference might be due to the physician's concerns about causing unnecessary stress to the parents of CWE and/or having a limited face-to-face interview time for each patient during their daily practice in the outpatient setting in Istanbul [27]. Of note, the main reason for this is out of the scope of this study and should be addressed in further studies.

Table 4
Comparison of epilepsy characteristics and degree of anxiety about dying from epilepsy.

		Worry about dying from epilepsy for CWE						
		Not at all		A little		Too much		
		n	%	n	%	n	%	
Duration of epilepsy	0–3 years	34	52.3	14	60.9	24	46.2	p =
	≥4	31	47.7	9	39.1	28	53.8	0.492
Severity of post-ictal confusion	Mild	36	56.3	7	31.8	7	13.5	p < 0.001
	Moderate	12	18.8	6	27.3	20	38.5	
	Severe	16	25.0	9	40.9	25	48.1	
Lifetime number of GTCS	0–5	41	67.2	17	73.9	20	38.5	
	≥6	8	13.1	3	13.0	16	30.8	p =
	Don't know	12	19.7	3	13.0	16	30.8	0.011
Current number of AEDs	0–2	60	93.8	18	78.3	37	71.2	p =
	≥3	4	6.3	5	21.7	15	28.8	0.005
History of seizures within the past year	Yes	43	66.2	18	78.3	43	82.7	p =
	No	22	33.8	5	21.7	9	17.3	0.113
	Yes	2	33.3	2	40.0	9	75.0	
My child is at risk for SUDEP	No	4	66.7	2	40.0	0	0	p =
	Don't know	0	0	1	20.0	3	25.0	0.018

Data are given in number and percentage, unless otherwise stated. SUDEP, Sudden unexpected death in epilepsy; CWE, children with epilepsy; AED, antiepileptic drug; GTCS, generalized tonic–clonic seizure.

During face-to-face interviews in the current study, most of the parents were willing to share their experiences about how they obtained the knowledge from their relatives and the way of coping with a CWE. We believe that close community relationships in our society may have affected the awareness degree and the way of coping with the disease in our study group, as patients and/or their relatives mostly tend to obtain information from their families and neighbors [28,29].

In two European studies, almost one-third of the patients obtained information about their disease through media or internet, and the use of internet was higher among the parents in a web-based survey, compared to those reached by a face-to-face interview [21–23]. Despite the growing interest on SUDEP in online platforms, we had only four parents who heard of SUDEP from internet, and only 13% of these parents were willing to be informed more specifically through online resources. This can be attributed to the limited number of web-based medical information resources in Turkish, despite the widespread access to internet in Turkey.

In the present study, most of the parents (87%) preferred visiting their treating physician to gain more information about SUDEP. Similarly, previous studies reported that the majority of patients and caregivers preferred obtaining information from physicians during a face-to-face interview [26,30–33]. In our study, over the two-thirds of the respondents also preferred to obtain more information about SUDEP, irrespective of the existing risk for SUDEP, and 54.7% preferred to obtain information at the time of the initial diagnosis. These findings are also consistent with previous studies on CWE [22,24]. In addition, similar findings were reported from the surveys including bereaved relatives of PWE who experienced a sudden death [32,33].

The present study mostly focused on the level and the need of knowledge of PWE, rather than their social and cultural beliefs. There might be additional social factors surrounding knowledge of SUDEP, such as religiosity and resorting to traditional healers. Chronic disease conditions, and in particular, epilepsy, may trigger to obtain epilepsy-related information and alternative treatment from traditional sources. Recent studies in Turkey found that people with epilepsy have had recourse to spiritual healing methods, such as, consulting a hodja, wearing amulets, and lead-pouring [34,35]. Although we added open-ended options to the questions to assess SUDEP prevention and risk reduction measures taken by parents, none of the parents mentioned such traditional methods. Accordingly, we probably failed to identify some of the participants lay beliefs about illness and the traditional healing methods that they used.

There is poor evidence on literature about relationship between the prior knowledge of SUDEP and seizure severity [26,36]. Although seizure severity is an important component of the overall severity of epilepsy, it does not capture other important aspects such as seizure frequency, medication needs and side effects, and seizure effects on daily life activities. Moreover, in the pediatric population, it is difficult to establish an objective scale of severity due to some methodological problems [37]. For example, restrictions originating from age or mental status or children and inadequate self-assessment of their epilepsy make the self-report scales impossible. Additionally, there is lack of evidence about the validated parent-based scales on severity of epilepsy in literature [38], and yet, there are on-going studies to clarify this issue [39]. Hence, we focused more on the risk factors related with SUDEP such as duration of epilepsy, frequency of seizures, and life-time number of GTCS. Prior knowledge of SUDEP was significantly associated with prolonged postictal confusion, epilepsy lasting more than three years, lifetime number of GTCS, and using three or more AEDs for seizure control. On the other hand, neither frequency of seizures nor parental education was found to be significantly associated with the prior knowledge of SUDEP. Since nocturnal monitoring for preventing seizures barely known in our study group, we did not include this data for comparison.

Our data shared the same results with a previous study concerning the factors causing anxiety about the death of epilepsy-related causes among parents [26]. The degree of anxiety about death of epilepsy-related cause among parents was significantly related with prolonged postictal confusion, lifetime history of having ≥6 GTCS and using three or more AEDs, but not significantly associated with the duration of epilepsy.

Parental worries about the death of epilepsy-related causes can be overcome by discussing risks of epilepsy and SUDEP. Although the guidelines recommend to inform patients and parents [3], discussing SUDEP has been an inconclusive topic for physicians.

Many studies in the European countries on epilepsy among children have shown that treating physicians are not most frequently willing to discuss SUDEP with patients and their parents. Underlying reasons for such behaviors among physicians may be related to hesitation of causing unnecessary anxiety, particularly for the parents of CWE who are at a low risk for SUDEP [40,41]. In our study, most of the parents (69.6%) who had prior knowledge of SUDEP were scared and anxious, when they first heard of it. The provision of SUDEP obtained from nonprofessional resources and lack of information about the associated risk factors may be the main reason for increased anxiety among parents of CWE. Thus, learning SUDEP for the first time from a physician or a healthcare professional may help patients or caregivers to overcome their worries. Nonetheless, further qualitative studies are needed to elucidate the main reasons behind the parental worries about the death of their children.

In addition, more than half (64.3%) of the parents of CWE reported that learning more about SUDEP could make a difference in how well they managed the disease of their children. A number of parents also believed that adherence to the treatment decreased the risks for SUDEP, and this ratio would likely increase with improved educational programs. Long-term parental behavior should be examined to analyze all aspects of perception of SUDEP.

Parents' coping capability and well-being may be influenced by culture and social belief. A wide variety of social qualitative studies have been carried out to assess epilepsy-related knowledge, needs, challenges, and perceptions of families [42,43]. However, data on SUDEP are scarce. Further qualitative studies may help us gain insight on how cultural convictions influence knowledge and management of SUDEP as well as understanding parents' coping mechanisms.

5. Strengths and limitations

The main strength of the present study is its relatively large sample size, a multicultural background of the Turkish population, and a broad range of CWE referring to our clinic, which provide a better understanding of the importance of knowledge on SUDEP among parents, and their attitudes and behaviors toward their CWE. Of note, each parent of CWE was consented in person, as face-to-face approach to the parents to prevent misunderstandings and inaccurate answers. In this way, they may trust to share their thoughts without prejudgment. However, social desirability and recall bias might occur to some degree, particularly regarding questions such as taking preventing measures and seizure characteristics.

Nonetheless, there are some limitations to this study. Because of the location of our hospital, the majority of the participants had lower socioeconomic status and education level compared to the general population of the city [44], which may effect results to represent the overall population. Also, certain subgroup analysis was limited by small numbers, resulting in some elements being under-powered to detect subtle differences. Furthermore, except options written in the survey, answers about methods that parents used to prevent seizures and SUDEP has not been systematically assessed. These answers may lead next studies to understand parental approach to seizures and coping mechanism. We enlighten certain aspects of this topic in Turkey, but there is an urgent need for studies assessing the physician knowledge and experiences about sharing SUDEP. Further systematic studies will help in the formation of a guideline.

6. Conclusions

In conclusion, our results suggest that knowledge about SUDEP among parents with CWE, particularly having severe seizures and using multiple AEDs, is poor. These results, however, might be a motivation for pediatric

neurologists who are concerned to facilitate a discussion with parents and/or caregivers about epilepsy-related death and SUDEP. Preparing evidence-based documents or guidelines for healthcare professionals may be useful to increase awareness, thus encouraging them to provide more information about SUDEP. The efforts of healthcare professionals for SUDEP discussion may reduce anxiety of parents with CWE, thereby, helping them to take necessary precautions to reduce the risk. Accordingly, a positive behavior for parents of CWE may be developed to reduce the risk factors for SUDEP. We believe that the present study, as a pioneer survey, paves the way for future researches on SUDEP and epilepsy-related deaths in Turkey.

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Declaration of competing interest

The authors have no conflict of interest to declare.

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